

Cementoossifying Fibroma of the Maxilla: a Case Report

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Summary

Cementoossifying fibroma of the maxilla is an uncommon tumor. Lesions with fibrous and osseous components include fibrous dysplasia (FD), ossifying fibroma (OF), cementoossifying fibroma (COF) and cementifying fibroma (CF). Fibro-osseous lesions other than FD seem to arise from the periodontal membrane.

We present a clinical case of a young woman referred for evaluation of a mass in the right cheek. The mass had first appeared 4 years ago and was growing larger inwards. She was treated with surgical resection via a Weber-Fergusson approach.

The physical examination revealed a maxillary enlargement and an inreoral lesion which had almost effaced the jugogingival groove. The teeth were moveable and displaced. Imaging studies demonstrated a soft tissue mass in the superior right maxilla which invaded the right maxillary sinus. The differential diagnosis included fibrous dysplasia, osteoid osteoma, osteoblastoma, chronic sclerosing osteomyelitis, ameloblastoma, squamous cell carcinoma of the maxillary sinus, calcifying epithelial odontogenic tumor (Pindborg tumor) and calcifying odontogenic cyst (Gorlin cyst). Histology established a cementoossifying fibroma.

In our case of a cementoossifying fibroma, the differential diagnosis based on clinical manifestations and conventional radiographic studies was controversial. Histologic interpretation was critical, and led to correct treatment.

Key words: *fibroosseous tumors, cementoossifying fibroma, maxilla, radiology, histology.*

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CASE REPORT
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Introduction

Cementoossifying fibroma (COF) is considered a benign osseous tumor, very closely related to other lesions such as fibrous dysplasia, cementifying periapical dysplasia or cemento-osseous florid dysplasia, however forming its own entity according to the 1992 classification of WHO (1). It is a bony tumor of the maxilla of possible odontogenic origin. It is believed to derive from the cells of the periodontal ligament (2-4). This is a layer of fibrous connective tissue surrounding the roots. It contains multipotential cells capable of forming cementum, lamellar bone and fibrous tissue. Under pathological conditions neoplasms containing any or all of the components may be produced (9). More aggressive lesions usually involve the maxillary antrum where extensive growth is unimpeded by anatomic obstacles. Because all cementum containing lesions are theoretically of periodontal membrane origin, maxillary sinus spread after origin from upper premolar or molar teeth is a distinct possibility (9). They manifest themselves as slow-growing, asymptomatic, intraosseous masses, most frequent in females aged between 35 and 40 years (7,8). Differential diagnosis should be performed, preferably with other fibro-osseous lesions of the maxilla such as fibrous dysplasia or osseous cementifying dysplasia (5,6). Although central COFs of the mandible are common, central COFs of the maxillary sinus are not; only a few have been reported in literature (9).

Case report

A 36-year-old woman was referred for evaluation of a mass in the right cheek (Figure 1). The patient stated that the mass had first appeared 4 years ago and had been gradually increasing in size ever since. The host had no complaint of pain, visual disturbances, dysphagia or dyspnea. Her past medical history was normal. The physical examination revealed maxillary enlargement and an intraoral lesion which had almost effaced jugogingival groove. The teeth were loose and displaced. Oral mucosa was normal. Imaging studies demonstrated a soft tissue mass in the superior right maxilla with expansive remodeling of bone and focal loss of cortical bone. The mass invaded maxillary sinus. It was well-circum-

scribed and showed radiolucent and radio-opaque features (Figure 2). CT scan showed a mixed density mass with diffuse scattered calcification involving the maxillary alveolar ridge, occupying and expanding the right maxillary sinus. A tissue sample was obtained for histopathological study and showed fibrous connective tissue with bone trabeculae and small, rounded, calcified foci that grouped into lobulated masses (Figure 3). COF was diagnosed, and patient underwent surgical resection via a Weber-Fergusson approach (Figure 4).

Discussion

Central cementoossifying fibromas are a distinct form of benign fibroosseous lesions of the mandible and maxilla. They are thought to arise from the periodontal ligament and are composed of varying amounts of cementum, bone, and fibrous tissue. Cementum is the mineralized connective tissue that covers the root of the teeth. The hybrid name central cementoossifying fibroma is used because there is a spectrum of fibroosseous lesions that arise from the periodontal ligament, ranging from those with only deposition of cementum to those with only deposition of bone (10). Central cementoossifying fibromas occur more frequently in women than in men. They arise in the mandible in 62% to 89% of patients, 77% occurring in the premolar region. Most are diagnosed between 20 and 40 years of age (10). When this tumor arises in children, it is called the juvenile aggressive cementoossifying fibroma, which presents at an earlier age and is more aggressive clinically and more vascular at pathologic examination (10). Central cementoossifying fibromas are asymptomatic until they cause expansion. Thus, they are generally not diagnosed until the tumor has had time to produce calcifications. Although central cementoossifying fibromas of the mandible are common, central cementoossifying fibromas of the maxillary sinus are unusual tumors. Central cementoossifying fibromas are typically well-circumscribed, solitary radiolucencies with scattered radioopaque foci. They maintain a spherical shape, expand the surrounding cortical bone without cortical perforation, and may cause tooth divergence. Large tumors may involve the nasal septum, orbital floor, and infraorbital foramen. The extent of the

tumor guides surgical therapy. Maxillary central cementoossifying fibromas are large at the time of presentation, indicating the capacity of the tumor to expand freely within the maxillary sinus. Pathologic examination of the central cementoossifying fibroma shows a proliferation of irregularly shaped calcifications within a hypercellular fibrous connective tissue stroma. The calcifications are extremely variable in appearance and represent various stages of bone and cementum deposition. Histologic differentiation between osteoid and cementum is difficult. In some cases, most of the calcified fragments are immature cementum, with basophilic coloration on hematoxylin and eosin-stained sections. These tumors are called central cementifying fibroma. In other cases, the calcified fragments are osteoid, with typical eosinophilic coloration on hematoxylin and eosin-stained sections. These tumors are called central ossifying fibromas. However, central ossifying fibromas can also be basophilic, causing difficulties in differentiating from central cementifying fibromas. Most pathologists feel that central cementifying fibromas and central ossifying fibromas arise from the same progenitor cell but produce variable amounts of bone and cementum within any one lesion. The hybrid central cementoossifying fibroma has evolved to indicate the likely presence of both types of tissue within the same lesion because of the difficulty in being able to distinguish reliably immature bone from immature cementum and because of the presence of both of these substances in many of the lesions. Thus, central cementoossifying fibroma is the most accurate histologic term, but it can be interchanged with either central ossifying fibroma or central cementifying fibroma. There is no apparent clinical or radiographic difference between the central cementifying fibroma or central ossifying fibroma, so the hybrid central cementoossifying fibroma also works well for radiography (10). Maxillary central cementoossifying fibromas tend to display a greater degree of immaturity than that seen in mandibular lesions, but there

is no reliable pattern useful to distinguish between maxillary and mandibular lesions. There is a correlation between the amount of calcification seen in the surgical specimen and that seen on the CT. The pathologic differences between central cementoossifying fibroma and fibrous dysplasia are few and the diagnosis must be made in light of the radiographic findings (10).

The differential diagnosis includes other lesions that contain radiopacities within a well-defined radiolucent mass: chondrosarcoma or osteosarcoma, fibrous dysplasia, odontogenic cysts, squamous cell carcinomas, calcifying odontogenic cysts (Gorlin cysts), and calcifying epithelial odontogenic tumors (Pindborg tumors). The well-defined border of the central cementoossifying fibroma helps differentiate it from aggressive sarcomas and carcinomas. Fibrous dysplasia has a characteristic „ground glass“ appearance, not seen in the central cementoossifying fibroma. The radiologic differentiation of central cementoossifying fibroma from Gorlin cysts and Pindborg tumors is difficult; the final diagnosis is based on histologic appearance. Pindborg tumors have a high association with impacted teeth (10).

The recommended treatment of the central cementoossifying fibroma is excision. The entire tumor should be removed including involved regions of the orbital floor and maxillary sinus walls. Central cementoossifying fibromas usually „shell out“ easily at surgery, but maxillary central cementoossifying fibromas are more difficult to remove completely than mandibular central cementoossifying fibromas. This may be attributable to the difference in bone character between the mandible and maxilla and to the available space for expansion in the maxillary sinus. Recurrence has been reported in as many as 28% of patients with mandibular central cementoossifying fibromas. The recurrence rate of maxillary central cementoossifying fibromas is unknown, but it is likely to be higher because of the greater difficulty of their surgical removal and larger size at the time of presentation (10, 11).